

Surgical treatment of odontogenic keratocyst by enucleation

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Abstract

Although odontogenic keratocysts (OKCs) are benign, they are often locally destructive and tend to recur after conservative surgical treatment. They must therefore be distinguished from other cysts of the jaw. Keratocysts possess outpouchings and microscopic daughter cysts from which recurrences may arise. Histologic examination is essential for diagnosis since the appearances on roentgenograms and at operation usually do not reveal the true nature of the lesion. Since many non-dental surgeons and pathologists are unaware of OKCs, a case is presented in which surgical treatment was by original conservative method. Decompression causes a reduction in the cyst volume with new bone formation so that the structures impinged upon (e.g., teeth, nerves) are completely free.

Keywords: Gorlin syndrome, odontogenic keratocyst, benign cystic neoplasm, radicular cyst

Introduction

The odontogenic keratocyst (OKC) is a cystic lesion of odontogenic origin, which is classified as a developmental cyst derived from the dental lamina. This lesion was first described in 1956 by Phillipson.^[1] It is one of the most aggressive odontogenic cysts of the oral cavity. OKC is known for its rapid growth and its tendency to invade the adjacent tissues including bone. It has a high recurrence rate and is associated with the basal cell nevus syndrome.^[2]

The distribution between sexes varies from equality to a male to female ratio of 1.6:1, except in children. OKCs may occur in any part of the upper and lower jaw, with the majority occurring in the mandible, most commonly in the angle of the mandible and ramus.^[2] The OKC involves approximately 11% of all cysts in the jaws and is most often located in the mandibular ramus and angle. This lesion can be associated, although not in all cases, with an impacted third molar. Radiographically, it appears as a unilocular or multilocular lesion with a scalloped contour.^[8] Radiographically, OKCs

demonstrate a well-defined unilocular or multilocular radiolucency with smooth and often corticated margins. OKCs tend to grow in an anteroposterior direction within the medullary cavity of the bone without causing obvious bone expansion. Displacement of teeth adjacent to the cyst occurs more frequently than resorption.^[10] OKCs of the maxilla are smaller in size compared to those of the mandible. When they are large, they tend to expand bone. No difference in site distribution was seen between unilocular and multilocular cysts.^[3] These lesions can also present as a small and oval radiolucency between teeth, simulating a lateral periodontal cyst. They can also appear as a radiolucency simulating radiographic presentation of the residual apical periodontal cyst.^[10]

Histologic features of these cysts were subsequently described by Shear and by Pindborg and Hansen. The keratocysts are classified as a developmental epithelial cyst and comprise approximately 11% of all cysts of the jaws. The age distribution appears to be bimodal. Ahlfors. Found a mean age of 41 years at the time of diagnosis in 255 patients. There appeared to be

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Figure 1: Facial asymmetry on right side of face



Figure 2: Large odontogenic keratocyst involving an impacted canine with unilocular and scalloped borders



Figure 3: Intraoral view showing obliteration of the labio-buccal sulcus



Figure 4: Degloving incision marked 6-8 cm away from attached gingival



Figure 5: Small perforation with exposure of impacted canine

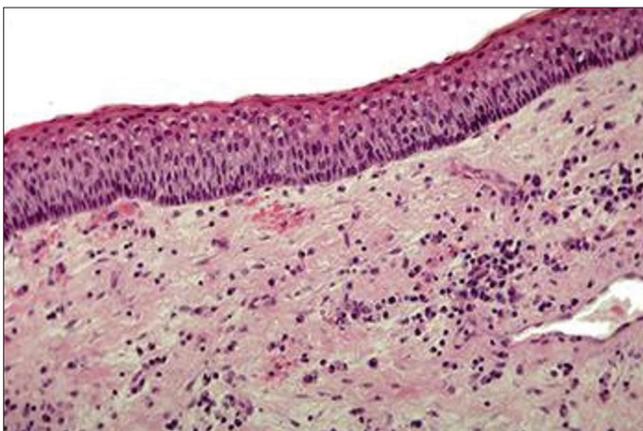


Figure 6: Composed of thin, irregular bundles of collagen, contains islands of epithelium that may represent daughter cyst

two incidence peaks between 25 and 34 years, and 55 and 64 years of age.^[11] The histologic features of OKCs include a thin epithelial lining, usually consisting of fewer than six cell layers



Figure 7: Lining was removed intact



Figure 8: Extraction of impacted canine

in a corrugated tissue composed of thin, irregular bundles of collagen, and often contain islands of epithelium that may represent daughter cysts. In many cysts, there is a tendency for the epithelium to separate from the underlying cyst wall.^[6] Histopathologically, they typically show a thin, friable wall, which is often difficult to enucleate from the bone in one piece, and have small satellite cysts within fibrous wall. Therefore, OKCs often tend to recur after treatment.^[1]

Cyst Contents (aspirate)

OKCs contain a dirty white, viscid suspension of keratin, which has an appearance of pus, but without an offensive smell. The following are used to test the presence of keratin.

- The smear should be stained and examined for keratin cells.
- Electrophoresis reveals low protein content, which is mostly albumin.
- Total protein is found to be below 4 g/100 ml.^[8]

Case Report

A 11-year-old female patient reported to the outpatient department of Modern Dental College and Research Center, Indore, in May 2010, with a complaint of swelling on the right side of the chin that caused considerable asymmetry of the face [Figure 1]. As narrated by her father, the swelling was noted 6 weeks previously, while its size increased gradually, but progressively. General physical examination ruled out the existence of a basal cell nevus syndrome. Intraoral examination was painless and caused no alteration in sensation over the mental nerve distribution. The labio buccal sulcus was obliterated [Figure 2] by a bony swelling and fluctuation was elicited in its center. Bony expansion extended from 72 to 46 lingually and the overlying mucosa showed normal texture. The



Figure 9: After curettage, betadine soaked gauze pack placed in the cavity

adjacent teeth were all mobile and were assigned grade II to grade III mobility. The radiographs taken were (ortho pantomogram (OPG), intra oral peri apical (IOPA), 12*7*6) and chest X-ray [Figure 2]. OPG revealed pathological resorption of the related deciduous teeth. (71,72,73,74,81,82,83) Fine needle aspiration cytology (FNAC) was done and pus was thick in consistency. Electrophoresis of the cystic fluid demonstrated low soluble protein content (<3.75 g/100 ml). Tentative diagnosis of OKC was made and confirmed later by histologic examination of the surgical specimen. The cross-section showed parakeratinized stratified squamous epithelium with uniform thickness in most areas and with a prominent palisading basal layer, which was consistent with the preoperative diagnosis of OKC. The operation was performed under general anesthesia by enucleation of the cyst, according to the principle of Partsch II.

Surgical procedure

Under all aseptic precautions in the operation theater, general anesthesia was given to the patient. Local anesthesia with adrenalin was given for tissue separation. Degloving incision was made 6-8 mm below the attached gingiva [Figure 4]. Mucoperiosteal flap was raised and then small perforation was made for aspiration of cystic contents and impacted canine was exposed [Figure 5]. Then, the cystic linings was removed and impacted teeth were removed [Figures 6 and 7] and curettage was done with carnoy's solution [Figure 9]. The site was closed with betadine soaked gauze pack and their borders are then sutured to create an open cavity that communicates with the oral cavity [Figure 8]. The specimen was sent for histopathologic examination and a diagnosis of OKC was revealed.

Discussion

The treatment of the OKC remains controversial. Treatments

are generally classified as conservative and aggressive. Conservative treatment generally includes simple enucleation, with or without curettage, using spoon curettes or marsupialization. Aggressive treatment generally includes peripheral ostectomy, chemical curettage with Carnoy's solution and resection. Some surgeons believe that the cyst can be properly treated with enucleation if the lesion is removed intact. However, complete removal of the OKC can be difficult because of the thin, friable epithelial lining, limited surgical access, skill and experience of the surgeon, cortical perforation, and the desire to preserve adjacent vital structures. The goals of treatment should involve eliminating the potential for recurrence while also minimizing the surgical morbidity. There is no consensus on adequacy of appropriate treatment of this lesion.^[9] Recurrence occurs due to the following reasons. The first reason involves incomplete removal of the original cyst's lining. Secondly, it involves growth of a new OKC from small satellite cysts of odontogenic epithelial rests left behind by the surgical treatment. The third reason involves the development of an unrelated OKC in an adjacent region of the jaws, which is interpreted as a recurrence. Marx and Stern believe that the two most common reasons for recurrence are incomplete cyst removal and new primary cyst formation.^[9]

The majority of cases of recurrence occur within the first 5 years after treatment. Because of the problematic nature of these cysts, many attempts have been made to reduce the high recurrence rate by improved surgical techniques. Bramley recommends the use of radical surgery with resection and bone transplantation.^[5] Decompression or marsupialization seem to be more conservative options in the treatment of OKC. Marsupialization was first described by Partsch in 1882 for the treatment of cystic lesions. This technique is based on the externalization of the cyst through the creation of a surgical window in the buccal mucosa and in the cystic wall. Their borders are then sutured to create an open cavity that communicates with the oral cavity. This procedure relieves pressure from the cystic fluid, allowing reduction of the cystic space and facilitating bone apposition to the cystic walls.^[1]

Currently, treatment involving careful and aggressive enucleation with close follow-up has been advocated for the OKC. John and James described the use of enucleation in conjunction with a chemical cauterizing agent and excision of overlying mucosa as a means of reducing recurrence.^[3]

Because the lining of the OKC is characteristically thin and friable, removal of the cysts in one piece may be difficult. Great care must therefore be taken to ensure complete removal of the cyst lining, without leaving behind remnants attached to the adjacent bone or soft tissue. The high recurrence rate associated with OKCs is a result of satellite

cysts confined to the fibrous walls of the OKCs. It should be emphasized that if the fibrous capsule is completely removed, no satellite cysts will be retained to serve as a nidus for recurrence. In view of the possible recurrence of the cysts from basal cell proliferation and because of the fragility of the cyst wall and the presence of satellite cysts, the osseous walls of the defect are abraded with coarse surgical or acrylic burs to ensure that residual peripheral cystic tissue is removed. Enucleation is not always easy because the lining may be extremely thin and friable, and access in the depths of the mandible may be limited. Multilocular cysts with bony trabeculae present special problems, in as much as it is difficult to remove the lining in one piece.^[6]

Enucleation with excision of the soft tissue overlying the OKCs has been proposed in an attempt to reduce the incidence of recurrence. A number of authors advocated the use of tanning with Carnoy's solution (absolute alcohol, chloroform, glacial acetic acid, and ferric chloride) before enucleation of the cysts. This procedure is often followed by excision of the overlying mucosa in continuity with the lesion.^[6]

Conclusion

The OKC has been the subject of much debate over the last 50 years with respect to its origin, its growth, and treatment modalities. The obvious advantages of our treatment techniques are as follows:

1. Eradication of the pathologic lesion,
2. Reduction of the potential for recurrence and
3. Preservation of the continuity of the mandible, thus maintaining jaw function and shape.

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